

Surgery in "Siamese" Twins: A Report of Three Sets of Conjoined Twins Treated Surgically *

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RELATIVELY few reports of conjoined twins, commonly known as Siamese twins, have appeared in the medical literature. In only a few of these cases have attempts been made to separate the twins. Many die in the immediate neonatal period before an operation can be done, others are so joined that operative separation is impossible. Survival of one twin after operation has been unusual. Even more unusual has been separation with survival of both twins. We have been able to find only nine previously reported instances of conjoined twins in which both survived surgical separation. The purpose of this presentation is to review briefly the literature and to report three sets of Siamese twins operated upon at the teaching hospital of the University of Tennessee College of Medicine in Memphis since January, 1953.

The origin of the term "Siamese twins," which is often used in this country as a synonym for conjoined twins, originated with Chang and Eng Bunker who were born near Bangkok in 1811 to a half Siamese-half Chinese mother, and a Chinese father. Luckhardt has contributed interesting historical and pathologic facts concerning these twins.⁸ They were known in the native home as the "Chinese twins" and it was not until they came to this country to join the P. T. Barnum side show that they were called the "Siamese Twins." They both married and lived for many years near Mt. Airy, North Carolina. Chang was the father

of ten children, Eng the father of nine. There were evidences during life that the twins were physiologically distinct entities. The sickness of one had no effect on the other, so that while one would be suffering from fever the pulse of the other would beat at its normal rate. Chang drank heavily, but Eng never felt any influence from the debauch of his brother—a seemingly conclusive proof that there was no free interchange in their circulation. The twins often quarreled, sometimes even coming to blows. During one of these quarrels, they applied to their physician to separate them surgically, but he declined to attempt it. Chang died quietly one night while Eng was sleeping. When Eng awakened and learned of this, he cried out to his wife "my last hour has come," and sank away. The autopsy revealed no cause of death in Eng. It did reveal, however, that the liver was a common organ and that colored material injected into the portal vein of one found its way into the portal circulation of the other. An operation would probably have been fatal to both if it had been attempted.

Conjoined twins vary from two symmetrical well developed individuals to those in which one "twin" is represented only by a portion of a body attached to a larger more completely developed host. Symmetrical conjoined twins may be classified according to the part of the bodies which are joined or shared.¹⁰ Anterior joining may be at the xiphoid, the sternum and costal cartilages, or the umbilicus: xiphopagus, thoracopagus or omphalopagus respec-

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tively. Cephalic fusion is the craniopagus. Posterior joining is usually at the pelvis; the pygopagus. A variant of the pelvic fusion is the lateral type: the ischiopagus. The bridge of fusion often contains an essential internal organ. Anterior fusions may have a common liver crossing the bridge, or a partially shared gastro-intestinal tract. Craniopagi may share dura and accompanying dural venous sinuses, or even brain tissue. In pygopagi, the anus or the urogenital organs may be shared; or with a sacral union the spinal cord may be single at the level of joining. When a vital organ is shared, surgical division into two viable organisms is impossible, though it may be possible to save one by giving him the shared vital organ. Surgical success is most likely when the union between the twins is only of skin, subcutaneous tissue and possibly cartilage.

There is no report in the medical literature of a successful separation of craniopagus twins with both twins surviving, though a recent report in the lay press indicates that this may have been done.³ The famed Brody twins nearly survived heroic surgery but because of common venous dural sinuses, one twin suffered from cerebral congestion and succumbed 34 days after operation.⁶ The second is still living and developing fairly well at last report.²

"In October, 1912, [ischiopagus] conjoined twins were successfully separated at the Military Families Hospital, Portsmouth, by a R.A.M.C. officer whose name cannot now be traced. One of the sisters of this pair is alive today, aged 41, and the scar of her operation can be seen on the lateral aspect of the hip. The other sister died of pneumonia at the age of four months, but there are no records to show whether or not she in fact had congenital heart disease."¹

An example of separation of asymmetric conjoined twins, with salvage of the more developed twin by giving it the organs which had been shared with its smaller parasite, has been reported by Spencer.¹³ The one twin was living at eight months

with "a complete gastro-intestinal tract with Meckel's diverticulum and a malrotation; two kidneys, normal to palpation, each draining into a separate bladder; two uteri with four tubes and four ovaries, and three vaginas. . . ."

Ochsner recently reported in a motion picture the successful separation of pygopagus twins. *Time Magazine* has reported a separation of twins joined at the base of the spines.⁴

Separations of xiphopagi with survival of both twins are recorded: The first operation of this kind on record seems to be one described by Konig (1690). In spite of the time which has elapsed since this operation was performed the case is so well documented that it seems quite authentic. The subject was a female xiphopagus, and the two components were connected by a slender, fleshy band just below the xiphoid process. The umbilical cord apparently arose from this structure at the time of birth. The connection consisted only of a sheath of skin enclosing a cartilaginous bar and a muscular and fibrous mass without large vessels or nerves. There was no communicating passage between the two peritoneal cavities. The operation was performed in infancy by means of a ligature which was placed around the connecting band and drawn tighter and tighter until the structure was greatly reduced in size when it was separated with a knife. Both twins lived.¹²

In 1927, Holm, in Minnesota, performed the second successful recorded separation of xiphopagus twins.⁷ He delivered the twins in a farmhouse one as a cephalic presentation, the other as a breech. Following delivery, it was noted that the tissue bridge connecting the two was twisted. Rotating the infants so that they were face to face untwisted the bridge. They were joined from the xiphoid to the umbilicus over an area about four inches (9 cm.) in length. One umbilicus was present and entered the lower portion of the attachment.

At the age of seven weeks, after fluoroscopic studies established that the gastro-intestinal tracts were separate, surgical separation was carried out. A common peritoneum was found at the site of fusion but no organs were involved. The abdominal hiatus created in each twin by the separation was closed under some tension. Both twins did well and were in good health at the time of the report nine years later.

In 1936, McLaren of the Colonial Medical Service successfully divided a native female xiphopagus in Nigeria.⁹ The case history is very similar to Holm's in that at birth one presented as a cephalic, the other as a breech; and the bridge of fusion was similar in extent and content. Operation was performed at the age of six months. "The stronger twin was anesthetized first in the hope that the other would go under, but after ten minutes of full anesthetization no obvious effect was observed, and the second twin had to be separately anesthetized." Both were doing well at the time of the report six weeks after the operation.

Reitman *et al.* in 1953 reported another very similar case of xiphoid joined female twins.¹¹ Surgical division was successfully performed on the day of delivery. The peritoneal cavities were entirely separate. Both were doing well at the age of nine months.

Dragstedt recently divided female xiphopagi who were natives of Siam and who were united in the upper abdomen in the same manner as the original Siamese twins.⁵ The tissue bridge contained a band of liver about three inches (7 cm.) in diameter, and there was a common peritoneal cavity. They have been adopted by an American woman and are developing normally.

Three pairs of conjoined twins have been operated upon on our service at the University of Tennessee College of Medicine, Memphis, since January, 1953.

Case 1. The "S" Twins. Xiphopagus type white males delivered on January 15, 1953, by Caesarian section eight months' gestation, at Philadelphia,

Mississippi, by Dr. Robert Bledsoe now of Greenville, Mississippi. They were transferred to the John Gaston Hospital on the following day. Combined weight on admission was 10 lbs, 11 oz. (4.84 Kg.). Examination revealed twins who were joined face to face from the manubrium to the umbilicus (Fig. 1). Below a common umbilical cord was an omphalocele measuring 7.5×15 cm. The color of the twins was good except for some cyanosis of the feet of one. Respirations were normal in character, and breath sounds clear. Heart tones normal. We felt that the omphalocele should be treated surgically after the method of Gross. A cannula was placed in the ankle vein, and operation was performed by Dr. J. D. Hardy. Skin was undermined surrounding the diamond-shaped omphalocele defect sufficiently to approximate the skin over the defect. The twins did fairly well for 2 days, but on the third day following operation both became cyanotic and died within 30 minutes of each other. At autopsy there was a common small intestine from the distal duodenum to the terminal ileum, and one twin had strictures at the uretero-pelvic junction bilaterally. It was thought that these abnormalities had not contributed to death however, and further that these abnormalities might have been compatible with life. The cardiovascular system was so anomalous as to be incompatible with life: there was a single inferior vena cava, and but two atria and three ventricles in the partially shared heart. It was felt that this was the cause of death, and would surely not have been amenable to surgical separation.

Case 2. Claudette and Constance M. A colored woman at term and in labor was admitted to the John Gaston Hospital on May 6, 1954. Auscultation revealed 2 fetal heart tones and an x-ray demonstrated brow united conjoined twins. Accordingly, Caesarian section was performed without difficulty. Combined birth weight was 10 lbs. $5\frac{1}{2}$ oz. (4.69 Kg.). Except for a brow to vertex union, the twins appeared normal in every respect and developed normally up to the time of surgical separation at age 5 months (Figs. 2 and 3). Pneumoencephalography prior to operation indicated that there were separate brains with a septum between, but with a small communication through the common septum. Intravenous methylene blue dye in one twin equilibrated with the second twin in less than 5 minutes, indicating a rather large vascular communication. However, when surgical anesthesia was induced in one twin, the second showed very little effect. One week prior to separation a delay of skin flaps in both was done so that a bucket-handle, temporally attached flap would be available to cover the anticipated defects

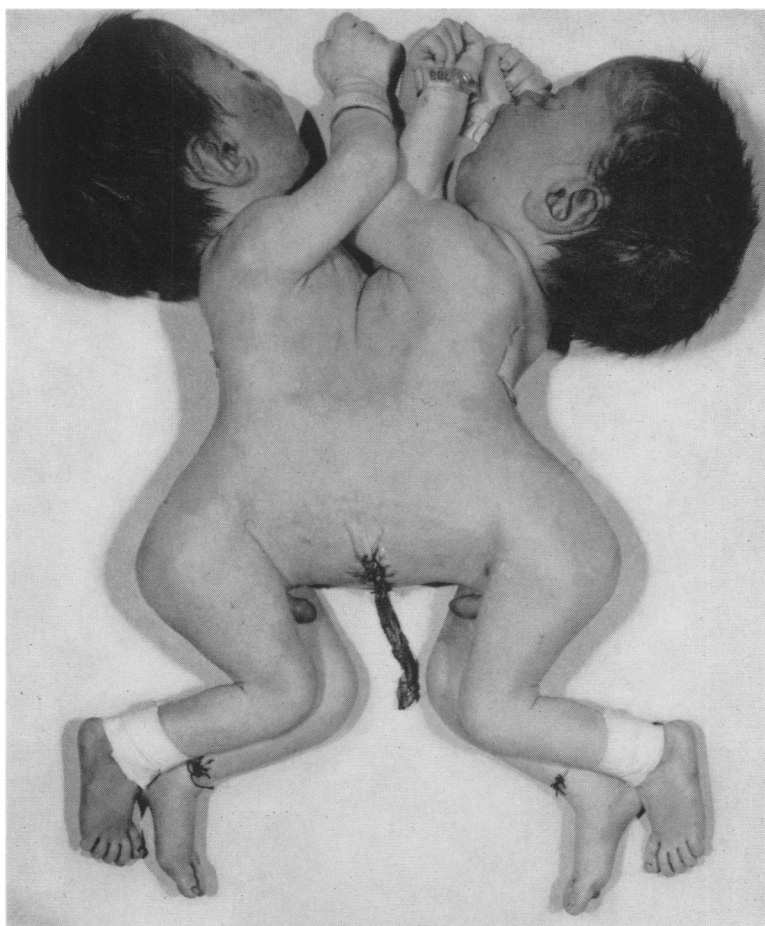


FIG. 1. The "S" Twins shortly after repair of omphalocele.

in the frontal region. The surgical separation was performed by Dr. Francis Murphey while the plastic procedures on the scalp were managed by Dr. Milton Adams. At operation, after removal of a wedge of frontal bone from each, a common dura was found with a central defect about 2.5 cm. in diameter. The twins shared the sagittal sinus, and there were also several smaller vascular communications. In order to separate the twins, it was necessary to divide the sagittal sinus in Constance as well as the inferior longitudinal sinus in this twin. Following operation, Constance appeared to be in shock which could not be reversed despite heroic therapy, and succumbed 5 hours later. Autopsy did not reveal any anatomic cause of death though there was some congestion of the dural vessels. Claudette did well and 5 days later the skin defect over the parietal and vertex regions created by shifting the flap forward was covered with an homologous skin graft taken from Constance shortly

after death. This graft "took" 100 per cent. Claudette has continued to develop normally, though she had to be hospitalized several times because of fluid accumulation at the separation site. An acrylic prothesis to close the bony defect in the frontal region was inserted on September 10, 1956. However, a draining sinus persists and revision will be necessary. Autogenous rib grafts will probably be used.

Case 3. *Linda and Lillian M.* Colored female conjoined twins were delivered vaginally without anesthesia at home on September 14, 1955, by Dr. C. C. Battle of Indianola, Mississippi. When two days old they were admitted to the John Gaston Hospital for evaluation and possibly surgical separation. Their combined weight on admission was 10 lbs., 2 oz. (4.59 Kg.). Physical examination revealed no abnormalities in either twin except for the bridge of tissue which joined them from the lower body of the sternum to the umbilicus (Fig.



FIG. 2. Claudette and Constance M. Craniopagus twins prior to surgical separation.

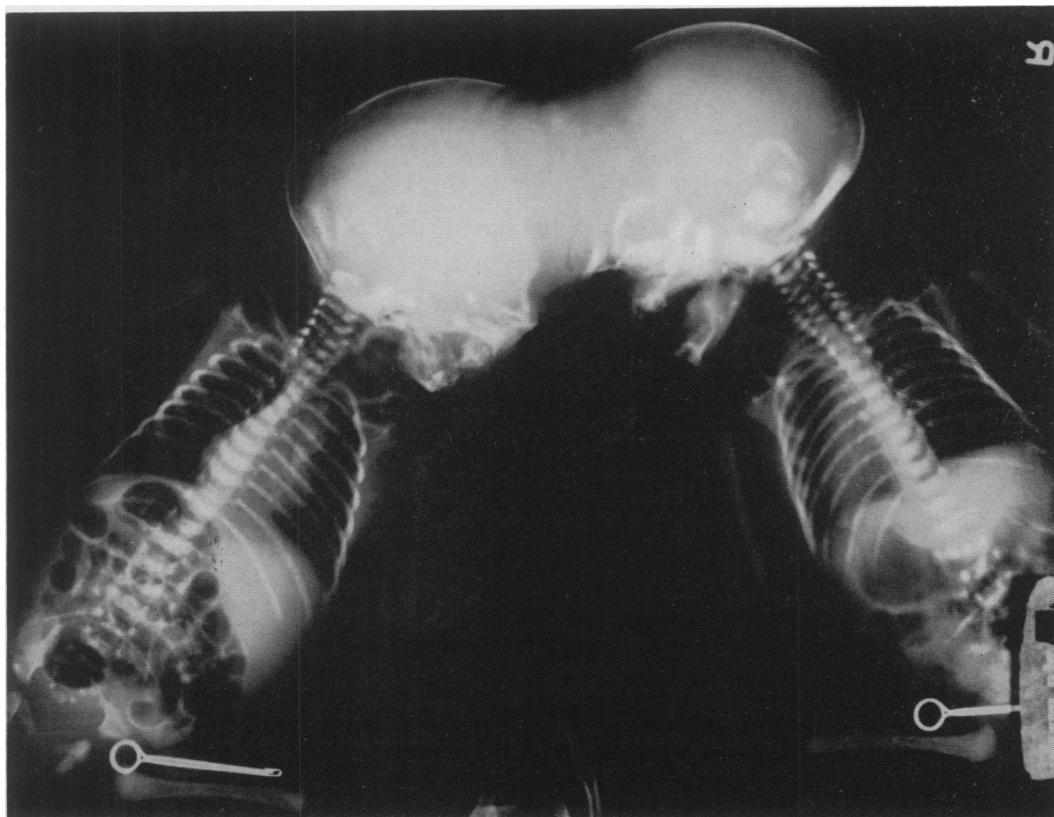


FIG. 3. Claudette and Constance M. X-ray demonstration of bony connection.

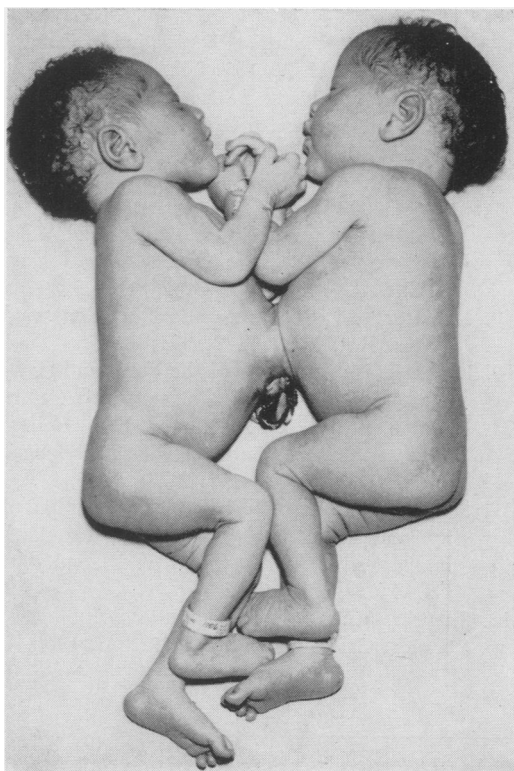


FIG. 4. Linda and Lillian M. Xiphopagus twins prior to surgical separation.

4). Their development prior to operation followed the normal pattern for twins of their size. X-rays taken after one twin had been given barium by mouth showed that the intestine of the one herniated across the bridge into the abdominal cavity of the other, but no communication between the twins' intestinal tracts could be demonstrated (Fig. 5). Also when charcoal was administered by mouth to one, it was recovered in the diaper of the one, but not in the other. Intravenous indigo carmine to one was almost all excreted in the urine of the one with only a faint tinging of the diaper of the other, indicating only a minimal vascular communication. Surgical separation was performed by one of us (H. W.) when the twins were $5\frac{1}{2}$ weeks old, using procaine locally supplemented by ether inhalations. The connecting bridge consisted of skin and fascia, cartilage joining the sterna, and a small bridge of liver which was divided between clamps. Centrally, a common peritoneal septum was present so that in separation Linda received the peritoneal covering which resulted in widely opening the peritoneal cavity of Lillian. Closure of the facial defects in both was accomplished under some tension. The resulting ventral herniae were



FIG. 5. Linda and Lillian M. Gastro-intestinal series in one twin, showing herniation of viscera into the abdominal cavity of the other, but no communication between the twins' intestinal tracts.

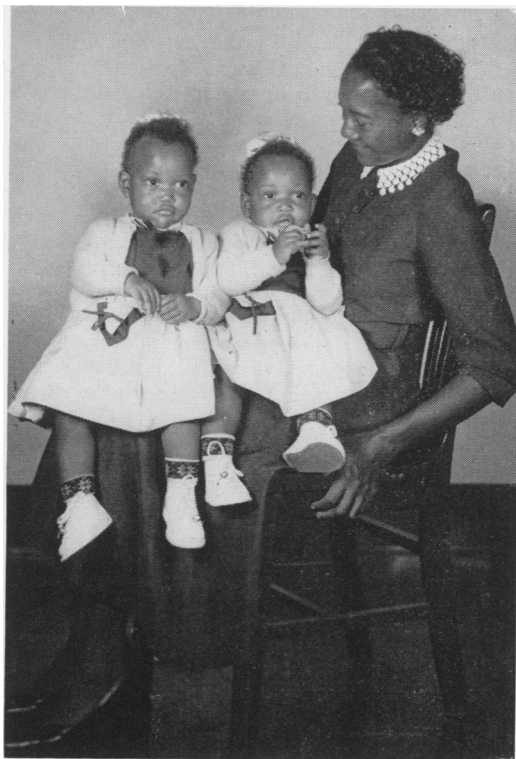


FIG. 6. Linda and Lillian M., age 14 mos., with mother.

repaired when the children were 4 months old. They have both done well and, when last seen at age 14 months, were entirely normal in every respect (Fig. 6).

SUMMARY

The various types of conjoined twins are discussed and a brief review of the literature concerning surgically treated cases is given. Three additional cases of conjoined twins operated upon since January, 1953, are reported.

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DISCUSSION.—DR. CLAUDE CRAIGHEAD, New Orleans, La.: This is to present the patients that Dr. Harwell Wilson alluded to in his paper. The twins were born on May 23, 1955, and have been reported in the May, 1956 issue of *Surgery* by Dr. Rowena Spencer.

(Slide) Here one sees the smaller twin on the right. This twin showed marked cyanosis, did not respond to painful stimuli, and the cyanosis ended at the trunk, indicating that there was possibly a common channel of the lower extremities.

(Slide) This shows a common union in the pelvis. This may be visualized by representing the pelvis as a closed 360° circle with the sacrum of the larger twin at 0°, and the smaller twin at 180°, with one symphysis at 90° and the other at 270°. The internal and external genitalia were in alignment with the pubic symphysis on either side.

(Slide) This shows the perineum and the external genitalia at right angles to the vertical axis of the twins. On one side there was a bipartite vagina, on the other side, a single vagina. On the side of the larger child was a crease but no opening. On the side of the smaller child was an anal opening.

(Slide) This shows the children at the time of operation. The operation had to be done immediately because the deteriorating situation in the smaller child was influencing the viability of

the larger child. The larger child was placed supine and given a general anesthetic.

(Slide) This is a diagrammatic representation of the findings encountered at surgery. There was one umbilical vein connecting from the single umbilicus to the liver of the larger child. There was a communication of about three centimeters of hepatic tissue between the livers. The small bowel of the larger child contained air. That of the smaller child contained no air, and apparently there was no communication with the upper gastro-intestinal tract of this child. At the confluence of the two ilei there was moderate dilatation of the terminal ileum. On the right is shown the anus on the smaller child's side. The bowel was divided at the terminal ileum where it joined the ileum of the larger child.

(Slide) This again shows the 360° arc previously alluded to; the sacrum of the larger child is at 0°, and the smaller child at 180°, with the symphyses at 90° and 270°. Each child contributed to each bladder one ureter, one tube to each uterus. The ureters of the smaller child were divided at each ureterovesical junction. The tubes and ovaries were left in.

(Slide) This was the end result in ten days.

(Slide) This is the perineum of the child with the rectum and the anus anteriorly, a crease posteriorly, a bipartite vagina on the left and a single vagina on the right. The child had two